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A rare case report of an adenomatoid odontogenic tumor associated with odontoma in the maxilla



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ABSTRACT

INTRODUCTION: The adenomatoid odontogenic tumor (AOT) associated with odontoma has been described as a new entity with overlapping features of adenomatoid-like structures and odontoma. Presentation of the case: We report the second case of AOT associated with odontoma in the posterior maxillary region. A 22-year-old patient presented expansion of the vestibular and palatine cortical wall of maxilla. Radiographic examination and computed tomography confirmed erosion of maxillary walls, root resorption, displacement of a neighboring tooth and radiopacities within the lesion. Histopathological analysis showed a cystic cavity lined by odontogenic epithelium, organized as duct like structures and tubular dentin. The diagnosis established was AOT associated with odontoma.

DISCUSSION: As fewer cases have been described, the aim of this study is to report clinical behavior and evolution of adenomatoid odontogenic tumor associated with odontoma, as it have not yet been described and its origin is not completely established. The hamartomatous or neoplastic origin of this tumor is under debate.

CONCLUSIONS: Therefore, it is necessary similar cases to be published to increase the knowledge about the clinical behavior and evolution of this tumor, to enable such lesions to be more clearly defined in the next classification of odontogenic tumors.

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1. Introduction

In the 1990s, the World Health Organization (1992) classified the adenomatoid odontogenic tumor as a benign tumor that presented odontogenic epithelium with odontogenic ectomesenchyme, with or without hard dental tissue formation. According to this classification, the hyaline material found inside the adenomatoid odontogenic tumor appeared to be a dysplastic dentin, and it occasionally showed a tubular pattern in some areas [1]. Thirteen years later, in 2005, a new World and Health Classification for Odontogenic Tumors was described in the book “Pathology & Genetics – Head and neck tumors” and the adenomatoid odontogenic tumor was reclassified as a tumor of the odontogenic epithelium with mature fibrous stroma, without odontogenic ectomesenchyme [2]. Here, the occurrence of a hyaline, dysplastic material or calcified osteodentin, containing (in very rare cases) dentinal tubules, was interpreted as a metaplastic process, due the absence of the odontogenic ectomesenchyme.

Many authors have reinforced the difficulty with classifying the adenomatoid odontogenic tumor based on its histogenetic origin, since it is an epithelial tumor and the “dentinoïd” material present is not a product of the odontogenic epithelium [3–5].

In the last decades, some cases of odontogenic adenomatoid tumor associated with odontoma have been described [3,4,6–11]. According to the literature review by Gomez et al. there are only eleven well-documented cases showing overlapping features of AOT and odontoma in the jaws, one of them involving the maxillary sinus [7]. The precise nature of this odontogenic tumor, whether it is neoplastic or hamartomatous, has not yet been completely established [3,4,6,9–11]. Consequently, some different terms have been adopted to define it, such as adenoameloblastic odontoma⁶, adenomatoid odontogenic tumor arising in an odontogenic cyst [7], adenomatoid dentinoma [3,9], adenomatoid odontogenic hamartoma [8] and more recently, adenomatoid odontogenic tumor associated with odontoma [4,10,11].

In addition, some authors have recommended applying the term adenomatoid odontogenic hamartoma when an attempt of tooth development occurs and fails [8].

In accordance with the SCARE criteria [12], we reported the second case of AOT associated with odontoma in maxilla presenting

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Fig. 1. Panoramic radiograph showing a well delimited radiolucency on the right side in the posterior region of the maxilla, absence of the first molar (16) and displacement of tooth 17 and 15.

aggressive behavior. The origin and nomenclature of such associated lesions was critically reviewed.

2. Presentation of the case

The patient, a 22-year-old woman was referred to the dental surgeon with the chief complaint of swelling in the right side of maxilla

and drainage of a yellow liquid into the mouth. The lesion was asymptomatic and clinical examination revealed facial asymmetry. Intraorally, expansion of the vestibular and palatine cortical bone of the maxilla were observed, covered with normal oral mucosa and without any signs of inflammation. Radiographically a unilocular and well-delimited radiolucency, in the right side of the maxilla was noted, with 8 months of evolution. Furthermore, displacement of the second maxillary right molar and root resorption of the teeth (second and third molars) involved were observed, as well as the absence of the first molar (Fig. 1). Cone-beam computed tomography revealed a hypodense lesion in relation to the maxillary sinus, from tooth 14–18 (Figs. 2 and 3 A). Axial and coronal views showed erosion of the vestibular and palatine sinus walls (Fig. 2). Root resorption of tooth 17 and 18 could be noted in the parasagittal view in the tomograph (Fig. 3B and C). Some radiopacities observed inside the lesion were suggestive of calcification or dental fragments (Fig. 3D and E). Fine needle aspiration (5 mL) of a citrine liquid and an incisional biopsy were performed. The clinical hypothesis was of odontogenic tumor or fibro-osseous lesion. Microscopic analysis showed a cystic cavity lined by a thin odontogenic epithelium forming cords or islands and with basal cells arranged in palisade, with vacuolated cytoplasm, while central cells resembled stellate reticulum (Fig. 4A and B). In some areas, the odontogenic epithelium was organized with duct-like structures

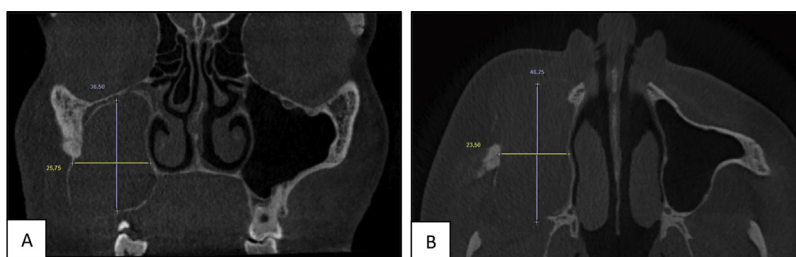


Fig. 2. Cone-beam computed tomography showing axial (A) and coronal (B) views, respectively, of the hypodense lesion and erosion of the vestibular and medial maxillary walls.

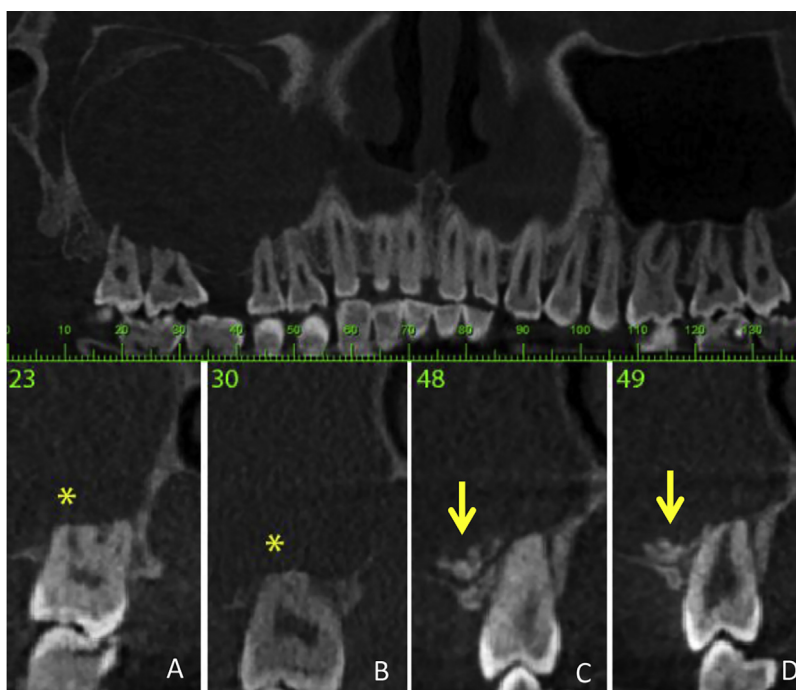


Fig. 3. Cone-beam computed tomography showing a unilocular well defined lesion in the posterior maxillary region (tooth 17, 18 and absent 16). A and B: Parasagittal views showing root resorption of the tooth 18 and 17 (*), respectively. C and D Some small radiopacities near the second premolar tooth (arrows), suggestive of dental fragments.

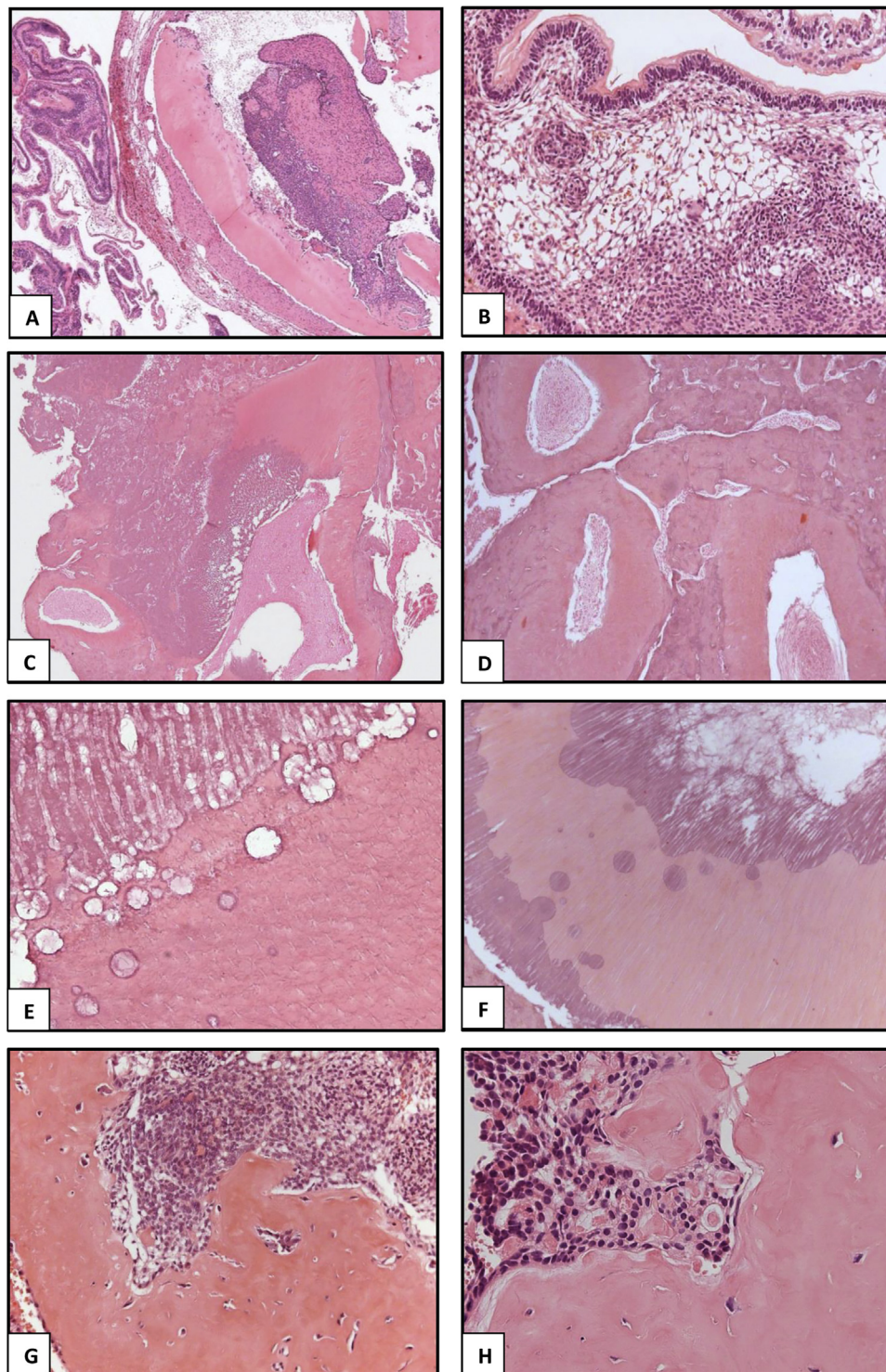


Fig. 4. A – Cystic cavity lined by a thin odontogenic epithelium forming cords or islands. Observe the dentinoid material in close contact with the odontogenic epithelium. B- Peripheral basal cells arranged in palisade, while central cells resembled stellate reticulum. C and D – Mineralized material containing dentin, suggestive of odontoma. E and F – Details of tubular dentin with globular calcification pattern, longitudinally cut. G and H – Osteodentin in close association with odontogenic epithelium.

containing an eosinophilic and amorphous material. Some foci of mineralized dentin characterized by a tubular formation and globular calcification (Fig. 4C–F) interlaced with osteodentin containing cells, were detected (Fig. 4G and H). The enamel matrix, predentin and ectomesenchyme were not evident in this tumor. According to the clinical, radiographic and microscopic features, the final diagnosis was of an adenomatoid odontogenic tumor associated with odontoma. The lesion was completely enucleated and during the

one-year follow-up period, there were no signs of recurrence, and new formed bone around the first molar tooth region was observed (Fig. 5).

3. Discussion

An adenomatoid odontogenic tumor associated with odontoma, such as in the present case report, has been described as a cys-



Fig. 5. Post-operative panoramic radiograph, one year after the surgery, showing new formed bone around the region of the first molar tooth (16) and no signs of recurrence.

tic lesion with overlapping features of AOT-like structures and an odontoma [3,4,6,10]. Most of the eleven cases described in the literature were indolent small intra-osseous lesions occurring in the posterior mandibular region, near the third molar tooth [3,4,6,8,9,11]. Furthermore, Tajima et al. described the first case involving the maxillary sinus, associated with the crown of a tooth, and aggressive behavior, such as expansion of the overlying bone.

Unlike those cases in the literature, the case we reported was the second large cystic tumor involving the maxillary sinus, but the first was not associated with a tooth in the maxilla. The radiographic exam contributed to initial detection of the lesion, but cone beam computed tomography was undoubtedly imperative to confirm the presence of small particles compatible with hard tissue inside the tumor, and the extension of the tumor margin. Moreover, the displacement and root resorption of neighboring teeth observed in our adenomatoid odontogenic tumor associated with odontoma were similar to features reported in other previously reported cases [3,7,9] reinforcing the neoplastic potential of this tumor.

Concerning the histopathological features, the present case showed areas of a typical adenomatoid odontogenic tumor pattern, such as a cystic cavity lined by a proliferative odontogenic epithelium in which the basal cells were arranged in palisade, with columnar polarized cells and inverted nuclei combined with duct like structures [2]. Mineralized material with tubular organization and globular calcification compatible with dentinoid was observed. This is a very specialized product characteristically of the odontogenic ectomesenchyme, but rarely seen in adenomatoid odontogenic tumors, according to the World Health Organization classification of tumors, 2005 [2]. Despite the dentinoid material, there was no evidence of predentin, ectomesenchyme and enamel matrix within our tumor, which reinforce the nomenclature adenomatoid odontogenic tumor associated with dentinoma suggested by other authors [3,8,9].

On the other hand, Vargas et al. reported two cases in which enamel matrix was unequivocally seen by electron microscopy, as well as dental papilla and tubular dentin, so they preferred to define these smaller intra-osseous lesions with limited growth and non-neoplastic potential, resembling a tooth in development, as adenomatoid odontogenic hamartomas.

Although the debatable nomenclature and origin of the AOT associated with odontoma [3,4,6–9,11], based on clinical, radiographic and microscopic features of the case reported, we emphasize that this tumor can occur as a benign tumor with aggressive neoplastic behavior, reaching large dimensions and with potential to cause bone and tooth resorption.

However, our doubt and that of other authors [4,6,9,10] remains, whether the adenomatoid odontogenic tumor associated with odontoma should be considered a variant of AOT or a distinct clinical- pathological condition.

4. Conclusion

Therefore, it is necessary for similar cases to be published to increase the knowledge about the clinical behavior and evolution of this tumor, to enable such lesions to be more clearly defined in the next classification of odontogenic tumors.

Conflict of interest

The authors declare no conflicts of interest.

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Ethical approval

The study doesn't require ethical approval.

Consent

The written informed consent has been obtained.

Author contribution

Assao, A. participated in the process of writing of the paper. Freitas, D.L. participated conducting the patient. Oliveira, D.T. participated of the process of writing the paper and of the microscopic analysis of the disease.

Guarantor

Dr. Denise Tostes Oliveira.

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